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Squamous cell carcinoma at a colostomy site

A.M. Ruiz-Beltran, S.A. Barquet-Muñoz, C.M. Sepulveda-Rivera, D. Cantu-de-Leon

Department of Gynecologic Oncology, Instituto Nacional de Cancerologia, Mexico City (Mexico)

Summary

Introduction: The development of a malignancy at a stoma site is an extremely rare event, with a rate reported at 2-4 of every 1,000 cases. Most of the cases are adenocarcinomas, with only a few squamous cell carcinomas (SCCs) reported as of today. *Case Report:* The authors present a case of a second primary SCC developing at the colostomy site nine months after colostomy formation. *Discussion:* The literature research revealed seven cases published in English. These cases presented with a minimum of ten years following stoma formation, and most of them developed in the setting of inflammatory bowel disease. It has been proposed that chronic chemical irritation at the stoma mucocutaneous junction may be carcinogenic. *Conclusion:* Patients with a stoma should be properly educated for early identification of problems. Additionally, a biopsy should be taken of any suspicious lesion in order to rule out malignant transformation.

Key words: Ovarian cancer; Stoma; Peristomal carcinoma; Squamous cell carcinoma; Second primary carcinoma.

Introduction

The development of a malignancy at a stoma site is an extremely rare event, with a rate reported at 2-4 of every 1,000 cases [1]. The majority of cases are adenocarcinomas, with only a few squamous cell carcinomas (SCCs) reported to date, particularly in patients with inflammatory bowel disease. It has been hypothesized that chronic irritation of the skin can result in a malignant degeneration.

The authors present a case of a second primary SCC developing at the colostomy site nine months after colostomy formation.

Case Report

A 60-year-old Hispanic woman, with a previous diagnosis of poorly differentiated endometriod ovarian carcinoma with right-ureter and sigmoid-colon involvement, who had a sub-optimal primary cytoreductive surgical procedure with a colostomy and right nephrectomy, was referred to the Instituto Nacional de Cancerologia (INCan) in Mexico City for further management of her disease three months after initial surgery. In the authors' institution, she was considered candidate to induction chemotherapy and completed six cycles of carboplatin-paclitaxel. A secondary cytoreductive surgical procedure was performed one month after finishing chemotherapy, without leaving any residual macroscopic disease.

During her follow-up, nine months later after her first surgery, the patient arrived with an indurated tender mass of about 1.5×1.5 cm arising from her colostomy. The patient referred noticing the mass during the past four months, having observed some intermittent bleeding. At the time, a biopsy was taken revealing an invasive, moderately differentiated SCC with lymphovascular space invasion (Figure 1). Immunohistochemistry was positive for p16 and p63, and negative for vimentin. Additional work-up studies, which included a CT scan of the patient's chest, abdomen, and pelvis, and a colonoscopy, did not show any metastasis. The case was further discussed at the institutional Tumor Board, and a three-dimensional (3D) tumor resection and colostomy remodeling was considered as the best option of treatment. A bowel resection was performed, revealing a 7×7×2.5-cm tumor without extension to skin borders and intestines free of neoplasia (Figure 2). Furthermore, the patient underwent external radiotherapy to her abdominal wall (30 Gy), demonstrating good clinical response. However, four months after the tumor resection, a chest CT revealed multiple bilateral lung lesions. A CT guided biopsy was compatible with poorly differentiated metastatic SCC. After that, palliative chemotherapy with carboplatin and gemcitabine was initiated, with the patient completing only two cycles because of neutropenic fever and septic shock development. She was able to recover and was referred to palliative-care team support. Finally, four months after discharge, the patient died at her home surrounded by her loved ones. Overall survival for ovarian cancer was 19 months, and nine months for SCC.

Discussion

SCC developing at a stoma site is a very unusual event. The literature research on revealed seven cases published in English literature [1-7]. These cases presented with a minimum of ten years following stoma formation, with the majority of these developing at the site of inflammatory bowel disease. The clinical summaries of all of the cases reviewed are presented in Table 1.

The exact mechanism of carcinogenesis at an ostomy site has not yet been not fully elucidated, although it has been proposed that chronic chemical irritation at the stoma's mu-

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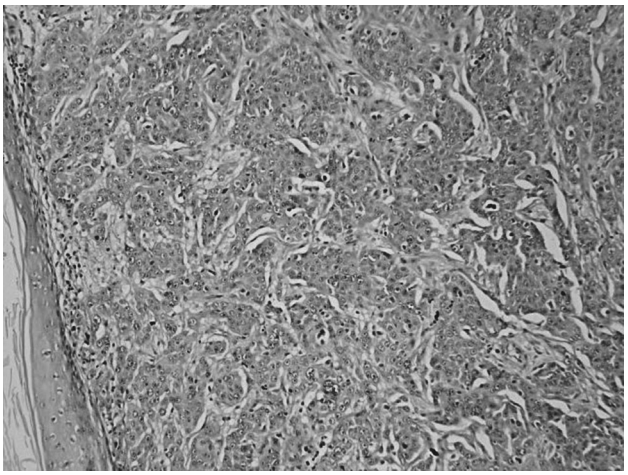

Figure 1. — Photomicrograph of invasive, moderately differentiated squamous cell carcinoma at colostomy site. Hematoxylin-eosin stain total magnification ×200.

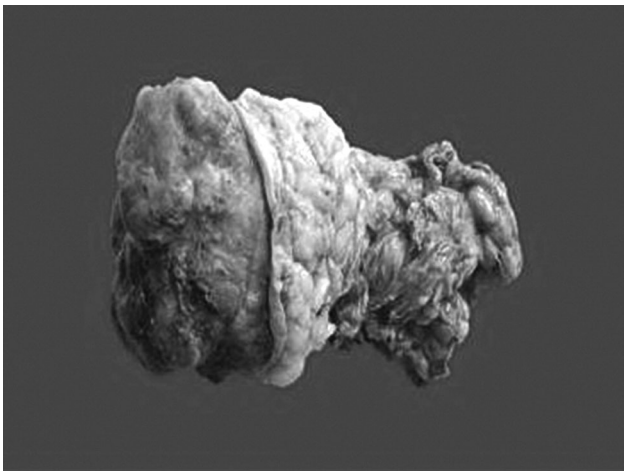

Figure 2. — Specimen showing showing the tumor widely excised with the stoma.

Table 1. — *Review of cases of squamous cell carcinoma at stoma site.*

Study	Age	Gender	Underlying diagnosis	Procedure	Time from primary surgery (yrs)	Pathology	Treatment
O'Connell *et al.* 1987	70	F	Crohn's colitis	Procto-colectomy and end ileostomy	26	Well differentiated SCC	Wide local excision and ileostomy relocation
Wu *et al.* 2000	67	M	UC	Procto-colectomy and end ileostomy	44	Well differentiated SCC	Wide excision and stoma translocation
Carne and Farmer 2001	66	M	UC	Procto-colectomy and end ileostomy	41	Moderately-well differentiated SCC	Wide excision and ileostomy relocation
Ramanujam and Venkatesh 2002	76	F	UC	Procto-colectomy and end ileostomy	51	Well differentiated squamous cell carcinoma	Wide excision and ileostomy relocation
Gupta *et al.* 2011	80	M	Transitional cell carcinoma	Radical cysto-prostatectomy and ileal conduit	27	Well differentiated SCC	Soft tissue radical resection and abdominal wall reconstruction
Ejtehadi *et al.* 2013	76	M	UC	Total colectomy and end ileostomy	54	Well differentiated SCC	Palliative radiotherapy
Reid *et al.* 2014	26	F	Neurogenic bladder	Monti ileovesicostomy	10	Moderately differentiated SC	En bloc resection of the stomal site and radical cystectomy
Ruiz-Beltran *et al.* 2016	60	F	Endometroid ovarian carcinoma	Sigmoidectomy and colostomy	<1 (9 months)	Moderately differentiated SCC	Wide excision and colostomy relocation

SCC = squamous cell carcinoma; UC = ulcerative colitis;

cocutaneous junction may be carcinogenic over a protracted period of time [8]. This malignant degeneration of a chronic inflammatory skin lesion is known as Marjolin's ulcer and is best described in chronic burn wounds. Chronic irritation and repeated attempts at healing provide a prolonged stimulus for cellular proliferation and may increase the rate of spontaneous mutations. However, carcinoma development is most likely multifactorial, affected by both environmental and genetic factors [9].

Clinical suspicion should be aroused with enhanced pain, malodorous exudates, bleeding, and vegetations. However, these should be confirmed with pathological interpretation of tissue biopsy specimens taken from multiple locations of the lesion and its margins to minimize false-negative findings.

Treatment should include wide excision with post-operative margin assessment with at least 4–6-mm negative macroscopic margins, and reconstruction techniques. In addition, adjuvant radiotherapy should be considered in the case of perineural involvement or positive tissue margins

[10].

In the case of the present patient, while there was irritation at the colostomy site, it was not long-standing, therefore, there must have been additional factors that contributed to carcinogenesis, such as previous primary carcinoma and systemic chemotherapy. Also, the present patient exhibited very poor disease progression despite adequate treatment. Marjolin's ulcer has previously been associated with poor prognosis and a high rate of metastasis.

Conclusion

Despite its being a very unusual event, there remains a small possibility of neoplasia development at an ostomy, and patients should be properly educated for early identification of problems. All patients should have good ostomy care in order to diminish the degree of chemical irritation at the mucocutaneous junction. Likewise, physicians should be aware of any changes in or complaints by the patients at each follow-up visit, particularly in patients with a previous primary neoplasia, because they are at a greater risk of developing a new carcinoma. A biopsy should be taken of any suspicious lesion to rule out malignant transformation.

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Corresponding Author:
S. BARQUET-MUÑOZ, M.D.
Instituto Nacional de Cancerologia
Avenida San Fernando #22, Colonia Seccion XVI
Mexico City 14080 (Mexico)
e-mail: sbarquet@gmail.com